Enhanced CPD DO C

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Multidisciplinary Management of Maxillary Regional Odontodysplasia

Abstract: This case report describes the multidisciplinary clinical management of a male patient, diagnosed at the age of 7, with Regional Odontodysplasia of the right maxilla. Following fixed orthodontic treatment to align his teeth, extraction of malformed teeth and subsequent bone grafting, the patient was successfully rehabilitated with implant-supported bridgework of a hybrid design to replace the teeth in the right maxillary quadrant. Clinical steps, as well as the complexities of this particular case, will be described.

CPD/Clinical Relevance: This case highlights the need for a multidisciplinary team approach to provide comprehensive care for patients with complex dental anomalies.

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Regional Odontodysplasia (RO) describes a rare localized developmental anomaly in which ectodermal and mesodermal tooth components are affected.[1] The term 'odontodysplasia' was first defined by Zegarelli *et al*[2] in 1963 to describe an uncommon dental developmental anomaly that affects both the deciduous and permanent dentitions. As the condition affects only one quadrant, Regional Odontodysplasia (RO) has become the accepted term. It tends to affect several adjacent teeth within a particular segment of the jaw and generally does not cross the midline. RO is normally located only in one arch with the maxilla being involved twice as often as the mandible and more frequently presents in the anterior region.[3-5] When primary teeth are affected, the permanent dentition is also usually affected.[6]

Characteristic clinical findings are yellow-brown discoloured teeth with an atypical morphology[7] that are soft on probing and often accompanied by gingivitis, swelling or abcesses. These teeth have an increased risk of caries and periapical inflammation and many fail to erupt.[3] Radiographically, the affected teeth show abnormal morphology and hypoplastic crowns.[5] The enamel and dentine are less radio-opaque than unaffected teeth, with a faint radiographic outline and a radiographic description of 'ghost teeth'.

Histologically, all structures of the dental germ are affected.[5] The enamel is hypoplastic, hypocalcified and may contain degenerated globular calcifications. The dentine is thin, tubules are reduced and clefts are often seen which create a direct communication between the pulp and oral cavity.[5]

Despite its well defined clinical, radiographic and histological features, RO is a condition of unknown aetiology. Although accepted to be non-hereditary, RO is more common in female than in male patients and there is no tendency towards a specific race or ethnic group. Several aetiological factors have been suggested in the development of this condition, including local vascular defects, latent viral infection of the tooth germ during development, metabolic or nutritional deficiencies and medications taken during pregnancy. However, no factors have been positively confirmed.[7]

The greatest difficulty in cases of RO arises in treatment planning and whether to remove the affected teeth. Several case reports have been described in the literature[9-14] relating to the management and restorative implications of this condition. Many argue a rationale for

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b

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Figure 1 (a–c) Teeth of the maxillary right quadrant severely malformed.

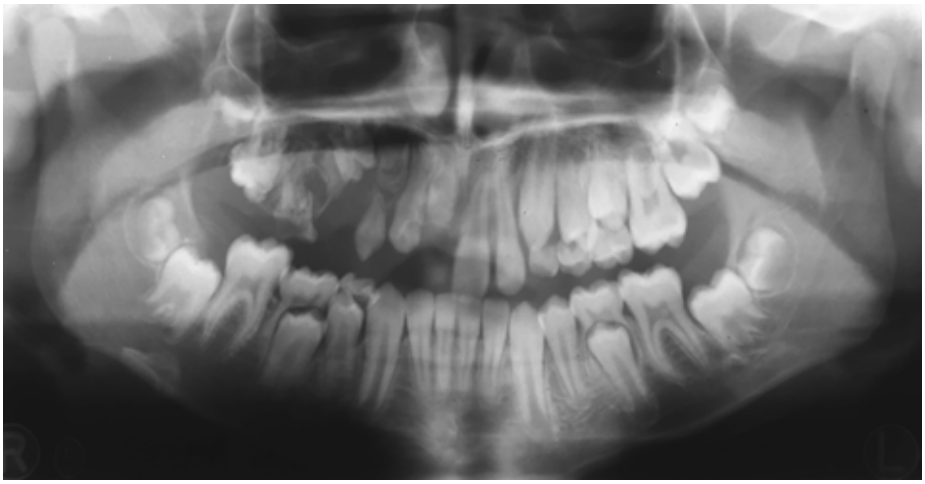

Figure 2. OPT of the patient taken at initial presentation (aged 7).

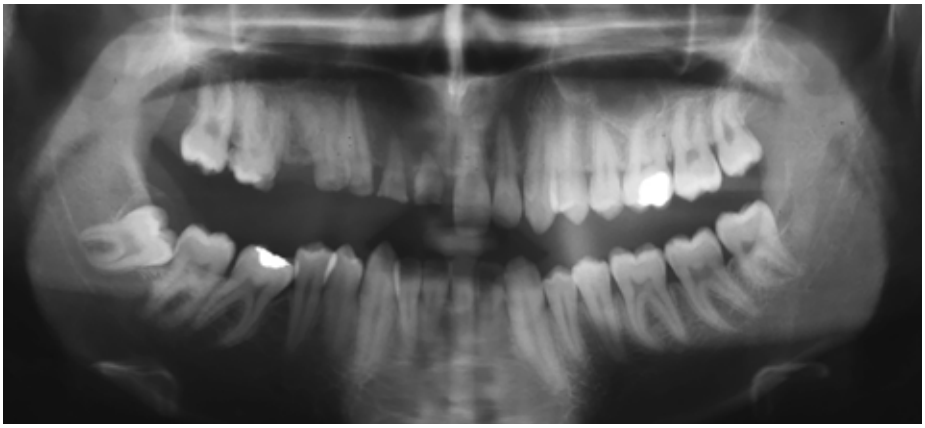

Figure 3. OPT taken in the permanent dentition.

the early extraction of teeth affected in Regional Odontodysplasia, as they are often unrestorable, may fail to erupt and risk development of dental abscess.[8,9] Contemporary thinking, however, has focused on conservative management, where feasible, in order to preserve these teeth for as long as possible and therefore support normal skeletal growth, jaw development and, importantly, to avoid the psychological effects of premature tooth loss.[4,6,9,10] Nevertheless, patient co-operation, condition of the teeth and patient/parent expectations have to be taken into account in an holistic approach to both preventive and interceptive philosophies.[4,6,10]

In the absence of symptoms and infection, case reports have demonstrated clinical and radiographic follow-up without any immediate management;[10] the provision of minimal restorations with acid etch composite;[8,9] or a fixed bridge prosthesis.[11] In more severely affected cases, recent literature has focused on multidisciplinary planning management[12] from a paediatric, orthodontic, oral surgery and prosthetic point of view to develop long-term options, including implant-retained prostheses[13] and removable prostheses.[4] This reports aims to discuss the multidisciplinary management of a severe case of RO.

Presentation

A 7-year-old male patient was referred to the Paediatric Department of Liverpool University Dental Hospital by a local orthodontist for 'consideration of the management of malformed teeth' in the upper right quadrant (Figure 1). Radiographic examination (Figures 2, 3) revealed severely malformed teeth, described as 'ghost teeth', in the maxillary right quadrant (UR6–UR1). As the patient was asymptomatic, a conservative management approach was adopted until the patient reached a suitable age for orthodontic intervention and significant growth had ceased, so that there could be a smooth transition from the orthodontic to the restorative phases of treatment and a long period of retention could be avoided.[14,15]

Orthodontic planning

At age 19, a joint orthodontic/restorative plan was devised with the aim of relieving the crowding, correcting the upper centreline and reducing the overbite to provide more vertical space in the maxillary right quadrant whilst establishing Class I incisor, canine and molar relationships.

Initially, treatment involved the use of a removable appliance to increase the vertical space between the upper and lower right quadrants. A bite plane in the lower right quadrant allowed over-eruption of the left buccal segments, resulting in an increase in vertical space between the right buccal segments where the malformed teeth were situated.

This was followed by the extraction of UL5, LR5, LL5 and the use of upper and lower pre-adjusted edgewise appliances (Figure 4) with an McLaughlin, Bennett and Trevisi (MBT) prescription, followed by upper and lower removable retainers. Once his

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Figure 4 (a–c) The use of upper and lower pre-adjusted edgewise appliances following MBT prescription once UL5, LR5, LL5 were removed.

Figure 5. Reconstructed CT scan of the affected maxilla prior to restorative treatment planning.

Figure 6. Surgical reconstruction of the right maxillary alveolus using an autogenous bone (ramus) block graft.

teeth were satisfactorily aligned, the patient moved into the prosthodontic phase of treatment to replace the malformed teeth with an implant-supported fixed bridge.

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b


Figure 7. (a, b) Post placement of four implants into the grafted right maxilla.

Figure 8. Try-in of a six-unit implant-retained superstructure prior to porcelain addition.

Figure 9. A customized verification jig to correctly orientate an angled abutment for UR3 fixture.

Prosthetic work-up

With an overriding prosthodontically driven treatment plan in mind, the prosthetic work-up began with the construction of a radiographic stent prior to Computed Tomography (CT) scanning of the maxilla (Figure 5), with the end goal of an implant-retained fixed bridge in the upper right quadrant. Scan results confirmed that the right maxilla was not suitable for immediate implant placement and would require bone augmentation to ensure implants were placed within a suitable prosthetic envelope.

At age 20, when any significant growth had ceased,[16,17] the malformed teeth (UR6–UR1, including the buried UR5) were extracted under local anaesthesia. After a suitable healing period, the patient underwent bony reconstruction of the right maxillary alveolus with bone blocks harvested from the right mandibular ramus (Figure 6), conducted under general anaesthesia.

Implant prosthesis

A surgical guide was then developed using a modified duplicate of the previously fabricated scanning prosthesis, for the placement of 4 Astra Tech OsseoSpeed™ (Astra Tech Dental, Stonehouse, UK) implants in the right maxilla after a 4-month healing period (Figure 7). A 2-stage (submerged) implant protocol was adopted and the implants were subsequently surgically exposed under local anaesthetic 3 months later. The definitive maxillary impression was made following mucosal healing and the mandibular fixed orthodontic appliance was subsequently removed just prior to the fabrication of the implant prosthesis, in order to maintain the vertical space which had been achieved by the orthodontic therapy.

The final six-unit superstructure (Figure 8) was developed as a screw-retained, metal-ceramic bridge, primarily for retrievability purposes. Due to the limitations of the angulation of UR3 abutment, a cemented unit included UR3 to maintain aesthetics. Astra 20° 'Uni' abutments were placed on UR6, 4, 1 fixtures and an angled, customized TiDesign™ abutment on UR3. Prosthetic management included the following stages:

- Secondary working impressions in polyether impression material;
- Occlusal registration and facebow transfer;
- Wax try-in of proposed clinical crowns;
- Metal superstructure try-in and methyl-methacrylate registration verification;
- Superstructure delivery with screw-retained components torqued to 15 Ncm at the time of luting UR3 unit with Tempbond® (Kerr UK Ltd, Peterborough).

A customized 'verification' jig was fabricated using addition-cured silicone and methyl-methacrylate, to

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Figure 10. (a–c) Final implant-retained fixed prosthesis *in situ*.

ensure successful orientation of the angled abutment UR3 (Figure 9). It was accepted from the outset that development of the natural papillae was unlikely; these were therefore obtained prosthetically with gingival shade combination of G24 and G23 porcelain to conceal remaining tissue deficiencies (Figures 10, 11).

Orthodontic retention, following the prosthodontic rehabilitation, was provided with maxillary and mandibular vacuum-formed retainers to be worn on a night-only basis. A tailored oral hygiene regimen with instruction/demonstration in the use of Superfloss® and interdental brushes in order to maintain peri-implant health was provided. Initially, the patient was reviewed clinically at short-time intervals, but these were extended as evidence of excellent plaque control was obtained. Radiographic review, 6 months post-loading, confirmed maintenance of peri-implant bone levels, although this will continue to be monitored in addition to any necessary supportive periodontal therapy.

Figure 11. A delighted patient with the final result.

Conclusions

Due to the distinct clinical, radiological and histological appearance of Regional Odontodysplasia, making a diagnosis is relatively straightforward. The difficulties arise in the management and treatment planning of these complex cases. Early decisions such as to whether or not to extract the affected teeth have to be made, and each case must be considered on an individual basis based on both motivational and clinical findings. In this case, intervention was delayed and the affected dentition in the upper right quadrant was maintained, until such time as a multidisciplinary solution could be provided.

This case highlights the need for a prosthodontically driven, multidisciplinary team approach to provide comprehensive care for patients with dental anomalies. The hybrid design allowed for optimal aesthetics, whilst retrievability of the structure was ensured. The patient received an excellent aesthetic and functional outcome that both delighted him and his family.

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